Primary pancreatic hydatid cyst: A case report and a brief review of the literature

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Abstract. Primary pancreatic hydatid cyst is a rare form of echinococcosis, even in endemic areas. The present study reports the case of a 67-year-old male patient with a primary pancreatic hydatid cyst who presented with severe epigastric pain, vomiting and fever for a period >2 weeks. An endoscopic ultrasound revealed a cystic lesion in the pancreatic head with a solid component. A computed tomography scan and magnetic resonance imaging confirmed the ultrasound finding. Under general anesthesia, the patient underwent total pancreatectomy and splenectomy. A histopathological examination confirmed a primary pancreatic hydatid cyst. Hydatid cysts rarely occur primarily in the pancreas. They may spread to the pancreas through the hematogenous route. Various procedures can be performed for cyst removal, depending on the size and location of the cysts. Open surgery, laparoscopy and hybrid options are available; however, to date, the gold standard is open surgery to prevent spillage and reduce the chance of recurrence. Although rare, primary pancreatic hydatid cysts can occur, with surgery being the main treatment modality due to the vague preoperative diagnosis based on imaging.

Introduction

Hydatid cyst, also known as cystic echinococcosis, is a parasitic infection caused by the *Echinococcus* parasite (1). It most commonly affects the lungs and liver, although other organs can also be involved (2). This disease poses a global threat as a parasitic infection, mainly affecting regions focused on farming and domestic animal husbandry. It remains endemic in several geographical areas, including South Africa, the Middle East, Australia, the Mediterranean and the Americas (3). There are four species of *Echinococcus*: *E. vogeli*, *E. granulosus*, *E. multilocularis*, and *E. oligarthrus* that can infect humans. However, the larval stage of *E. granulosus* is the cause of hydatid cysts in 95% of cases (4,5). Humans are incidental hosts to the parasite, becoming infected through direct contact with the definitive hosts (canines) or by ingesting food or water contaminated with the eggs of the parasite (6,7). Pancreatic hydatid cyst (PHC) is a rare form of hydatid disease, even in endemic regions where agriculture and stockbreeding are common occupations (8). The infection is commonly asymptomatic; however, signs and symptoms of PHC can include intermittent fever with shivering, loss of appetite, nausea, vomiting, epigastric pain, obstructive jaundice, weight loss and recurrent acute pancreatitis (9,10).

The present study reports study the a case of a 67-year-old male patient with primary PHC. The report follows the SCARE guidelines, and all references have been screened for reliability (11,12).

Case report

Patient information. A 67-year-old male from a rural area in the central-eastern part of Iraq visited the Internal Medicine Clinic at Smart Health Tower (Sulaymaniyah, Iraq) with severe epigastric pain, fever and vomiting for a period >2 weeks. The patient had a previous medical history of diabetes mellitus and hypertension. He had a history of cholecystectomy for gallstones 30 years prior. During this procedure, a bile duct stone was found, leading to the decision to perform a Roux-en-Y hepaticojejunostomy, since endoscopic retrograde cholangiopancreatography (ERCP) was not accessible in the region at that time. The patient also reported a history of contact with

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sheep and goats, as his family owned livestock. However, the history of ingesting contaminated foods was unclear.

Clinical findings. A physical examination revealed epigastric fullness and tenderness without jaundice. Laboratory investigations revealed a hemoglobin level of 13.1 g/dl (normal range, 13-17 g/dl), a white blood cell count of 21,600 mm^3 (normal range, 4,000-11,000 mm^3) and a platelet count of 531,000 mm^3 (normal range, 150,000-400,000 mm^3). Other biochemical analyses yielded results which were within the normal range, including a lipase level of 66.1 IU/l (normal range, 13-85 IU/l), a total serum bilirubin level of 0.462 mg/dl (normal range, 0.3-1.2 mg/dl), an alkaline phosphatase level of 111 IU/l (normal range, 40-130 IU/l) and an alanine aminotransferase level of 6.9 IU/l (normal level, <50 IU/l).

Diagnostic assessment. An endoscopic ultrasound (EUS) revealed a cystic lesion in the pancreatic head with a solid component. A subsequent computed tomography (CT) scan confirmed this finding. Magnetic resonance imaging (MRI) of the abdomen further detailed a multiloculated cyst situated in the head and uncinate process of the pancreas, measuring 54x58x51 mm. It had a thick enhancing wall with multiple thick enhancing septae, diffusion restriction, and no communication with the pancreatic duct (Fig. 1). At least eight local reactive lymph nodes were identified (all <7 mm in short-axis diameter). The lesion adjoined the superior mesenteric vein and portal vein for a distance of >5 cm; however, there were no signs of invasion into the gastroduodenal artery, hepatic artery, or superior mesenteric artery. Another cyst, measuring 20x20 mm, was observed at the posterior aspect of the pancreatic body, with an apparent connection to the side branch of the pancreatic duct. The common bile duct had a diameter of 10 mm, and the main pancreatic duct was 5 mm, both of which entered the mass region. There were, additionally, several small pancreatic cysts in the background that reached out to the spleen. Fine-needle aspiration cytology revealed a pyogenic inflammatory process with extensive pancreatic fibrosis and no evidence of malignancy.

Therapeutic intervention. Under general anesthesia and following the administration of a dose of 1 g Ceftriaxone intravenously supplied by Acino Swiss, an incision was made in the left subcostal extension. The intraoperative assessment revealed a large cystic tumor occupying the head and uncinate process, along with other cysts distributed over the pancreatic body. Total pancreatectomy with partial excision of the attached second portion of the duodenum and splenectomy were performed due to the presence of a large mass, as indicated by imaging findings. The mass nearly covered the pancreas and extended toward the spleen. None of the imaging findings suggested features indicative of a hydatid cyst, raising concerns among the surgeons and necessitating consideration of the aforementioned intervention. Hemostasis was achieved, and two corrugated drains were left in place, followed by thorough irrigation and layered skin closure. Of note, three biopsy fragments were sent for a histopathological examination: The duodenum with the attached head of the pancreas (16.5x8x5.5 cm), the spleen (11x7.5x3.5 cm) and an irregular gray-brown fragment of pancreatic tissue (11.5x5.5x2.5 cm). The sections (5-μm-thick) were paraffin-embedded and fixed with 10% neutral-buffered formalin at room temperature for 24 h. The sections were then stained with hematoxylin and eosin (Bio Optica Co.) for 1-2 min at room temperature and were examined under a light microscope (Leica Microsystems GmbH). The pancreatic head and the separate pancreatic fragment had a firm, dull, gray-cut surface with scattered white areas. A microscopic examination revealed chronic xanthogranulomatous pancreatitis, consistent with rare acellular fragments of the hydatid cyst wall and remnants of hooklets from the brood capsules (Fig. 2). It was consistent with a reaction to a hydatid cyst. All the peripancreatic lymph nodes were benign, and the spleen contained a benign spindle cell tumor, consistent with an inflammatory pseudotumor.

Follow-up. Following the surgery, the patient was admitted to the intensive care unit for 2 days. On the 4th post-operative day, the patient developed tachypnea and hypoxia. Consequently, a CT pulmonary angiography was performed, revealing a bilateral pulmonary artery embolism. He received 6,000 units of heparin intravenously in two doses, leading to an improved condition. He was discharged without complications on the 10th post-operative day. The patient received a 4-week course of albendazole at a dosage of 800 mg/day (PHARMA développement). The case remained stable without complications during the 6-month follow-up period, after which he was lost to follow-up. At 11 months after the surgery, his family reported his death due to increased insulin requirements and uncontrollable diabetes.

Discussion

Echinococcosis is a parasitic infection that originates from the *Echinococcus* parasite. It can affect almost every organ and body part (1,3). The primary hosts for the parasite are dogs, jackals and wolves, and the intermediate hosts are cattle, sheep, horses and humans (4). The liver and lungs are the most commonly affected organs, accounting for 90% of cases. The involvement of the pancreas is uncommon (1,3,9). The ingestion of food and water contaminated by the eggs of the parasite or direct contact with the primary host leads to infection (13).

Individuals who own livestock have a 3-fold greater risk of being diagnosed with *Echinococcus* infection compared to those who do not own livestock (14). The patient in the present case report owned livestock and had contact with sheep and goats. PHCs are uncommon, with a frequency ranging from 0.14 to 2%. PHCs are commonly unrecognized (90-91%) and distributed unevenly over the tail (16-19%), body (24-34%) and head (50-58%). In the case described herein, the PHC affected the head of the pancreas. The infection may reach the pancreas via the biliary ducts, retroperitoneal extension, lymphatic spread from the intestinal mucosa and hematogenous transmission through the pancreatic vessels (9). The majority of reported cases fall within 18-38 years of age (9), although the patient in the present study was 67 years of age. Several similar cases have been reported in the literature (4,8,9,15-23) and are summarized in Table I.

Pancreatic cysts grow over time (0.3-2 cm per year), and some patients remain asymptomatic for years before being

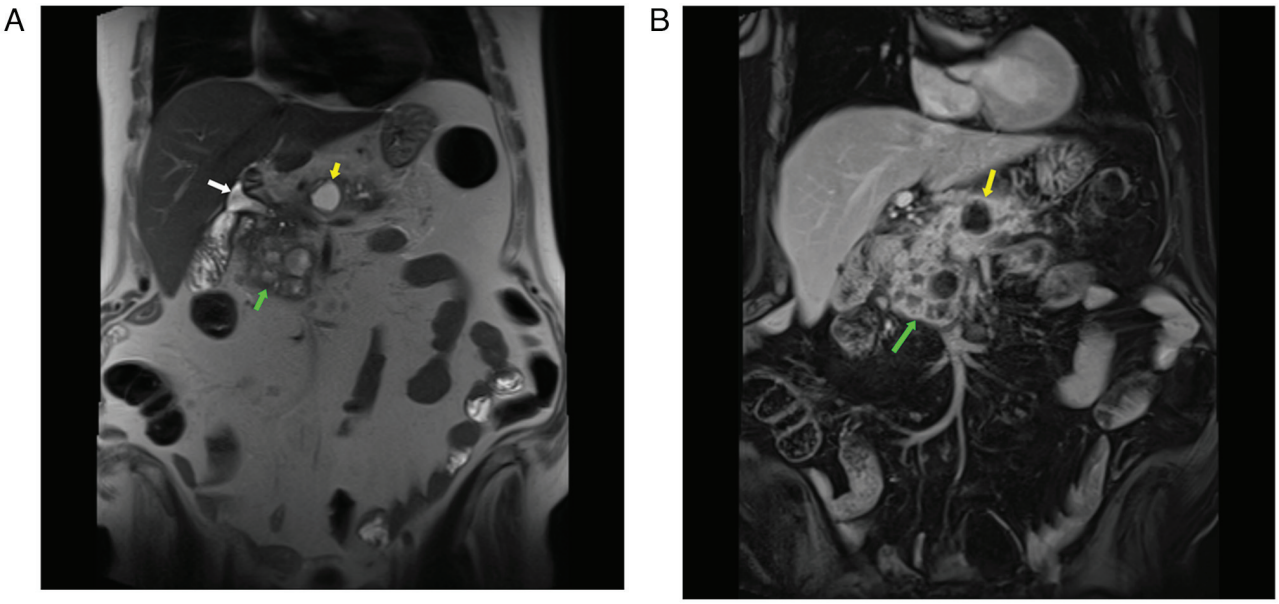

Figure 1. (A) Coronal T2 MRI image illustrates a multiloculated cystic lesion in the head of the pancreas (green arrow), a unilocular cyst in the body of the pancreas (yellow arrow) and common bile duct dilatation (white arrow). (B) Coronal T1 fat suppression post-contrast MRI image illustrating a multiloculated cystic lesion in the head of the pancreas (green arrow) and a unilocular cyst in the body of the pancreas (yellow arrow).

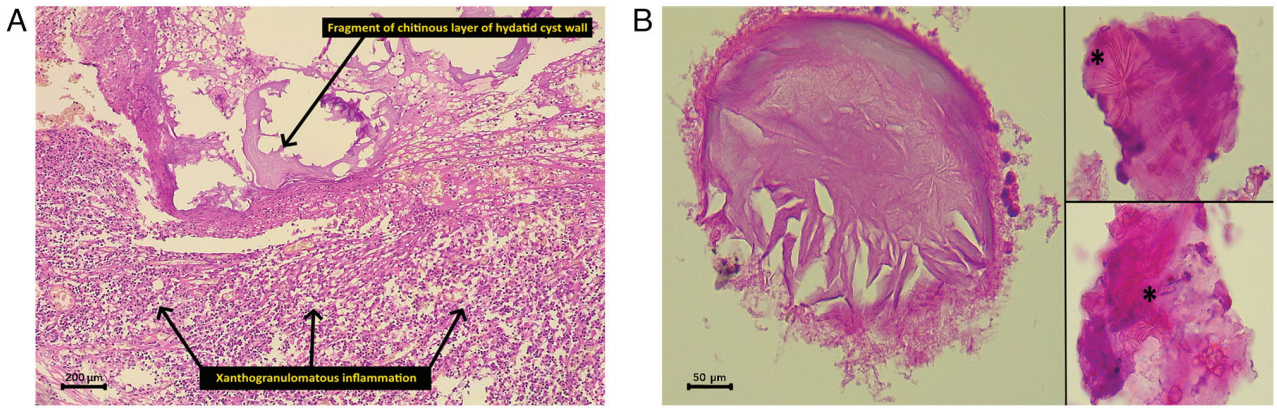

Figure 2. (A) Xanthogranulomatous inflammation, comprising mixed inflammatory cells with a predominance of foamy macrophages, surrounds an acellular, lamellar, eosinophilic fragment of the chitinous layer of the hydatid cyst wall (hematoxylin and eosin staining; magnification, x100; scale bar, 200 μm). (B) A collage of three different areas from the histologic sections of the pancreas containing acellular, eosinophilic fragments from the chitinous layer of the hydatid cyst wall in addition to remnants of hooklets (asterisks) from the brood capsules (hematoxylin and eosin staining; magnification, x400; scale bar, 50 μm).

diagnosed. In symptomatic PHC cases, the location of the cyst inside the pancreas determines the clinical findings and complications. The most commonly reported signs and symptoms are epigastric pain, fever, nausea, vomiting, weight loss and abdominal fullness (9). However, obstructive jaundice can develop if the lesion is situated in the head of the pancreas (22). The patient described herein presented with severe epigastric pain, fever and vomiting. He had no signs of jaundice due to the biliary tree bypass created by the hepatico-jejunostomy performed 30 years prior due to a bile duct stone. The World Health Organization Informal Working Group on Echinococcosis (WHO-IGWE) classification categorizes cysts into active, transitional and inactive, which is relevant for treatment planning and follow-up. In this classification, CE1 and CE2 are active cysts; the CE3 group represents transitional cysts, with CE3b being biologically active; and the CE4 and CE5 groups are inactive, late-stage cysts (24). In the case in the present study, imaging did not reveal any characteristics indicative of a hydatid cyst. Instead, there were signs of pancreatitis and the formation of an abscess. For that reason, it was inappropriate to apply the WHO classification to this case.

Direct hemagglutination, immune electrophoresis, skin tests and enzyme-linked immunoassays can be used to support the diagnosis; however, the validity of these tests is undermined by false-positive and false-negative results (17). Various imaging modalities are useful in the diagnosis of pancreatic cysts, including CT, MRI and ultrasonography (9). In the case in the present study, an MRI revealed a multilocular cystic lesion in the head and the uncinate process of the pancreas with a thick enhancing wall and multiple thick enhancing septae. EUS and ERCP can be utilized for aspiration or biopsy

Table I. Summary of some cases of pancreatic hydatid cyst in the literature.

Authors (year of publication)	No. of cases	Age (years)	Sex (M/F)	Symptoms	PMH	Cyst location on the pancreas	Cyst size (cm)	Management	POAR	Complications	Follow-up	(Refs.)
Kothiya *et al* (2022)	1	20	F	Epigastric Pain	None	Pancreatic head	6.46	Laparoscopic partial cystopericystectomy and omentoplasty	Albendazole 15 mg/kg/day, for 8 weeks	None	Uneventful	(15)
Cherouaqi *et al* (2021)	1	54	F	Epigastric and left hypochondriac pain	Hypothyroidism	Pancreatic isthmus	5	Surgical exploration and resection	Albendazole 800 mg/day, for three months	None	Uneventful	(16)
Alsaid *et al* (2018)	1	34	M	Abdominal pain, dyspnea, fatigue, and weakness	Acute pancreatitis	The space between the tail of the pancreas, spleen, left colic angle, left kidney, stomach, and diaphragm	21.8	Surgical exploration and cyst fenestration	None	Acute edematous pancreatitis and deep vein thrombosis	Uneventful	(4)
Ozsay *et al* (2018)	1	23	F	Epigastric pain	None	Pancreatic tail	4.47	Surgical exploration and enbloc resection of distal pancreas and spleen	Albendazole 800 mg/day, for three months	None	Uneventful	(17)
El Bakkaly *et al* (2017)	1	5	F	Epigastric pain, dietary vomiting, and diarrhea	None	Pancreatic head	N/A	Surgical exploration, cyst sterilization, and resection	Albendazole for 6 months	None	Uneventful	(18)
Ahmed *et al* (2016)	1	40	F	Epigastric pain	None	Pancreatic head and tail	5.4	Surgical exploration, cyst sterilization, and partial cystectomy	albendazole 10 mg/kg/day, for 8 weeks	None	Uneventful	(8)
Makni *et al* (2012)	1	38	M	Abdominal pain, nausea, and vomiting	None	Pancreatic corpus	9.4	Surgical exploration, dissection of the pancreatic tail, and distal pancreatectomy with splenectomy	Albendazole 800 mg/day, for three months	None	Uneventful	(9)

Table I. Continued.

Authors (year of publication)	No. of cases	Age (years)	Sex (M/F)	Symptoms	PMH	Cyst location on the pancreas	Cyst size (cm)	Management	POAR	Complications	Follow-up	(Refs.)
Suryawanshi *et al* (2011)	1	20	M	Epigastric pain and occasional vomiting	None	Pancreatic head	8	Laparoscopic cyst evacuation and omentoplasty	Albendazole 10 mg/kg/day, for 3 months	None	Uneventful	(19)
Derbel *et al* (2010)	7	25	F	Left hypochondriac pain	N/A (all)	Pancreatic tail	6	Laparotomy and cystectomy	N/A (all)	None	Uneventful (all)	(20)
		19	F	Right hypochondriac pain		Pancreatic tail	7					
						Pancreatic body and tail		Laparotomy and cystectomy		None		
		32	F	Epigastric pain Left hypochondriac pain and fever		N/A	15	Laparotomy and cystectomy		None		
		41	M	Epigastric pain and jaundice			15			None		
				Epigastric pain and jaundice		Pancreatic head		Laparotomy, cystectomy and splenectomy				
		38	M	Left hypochondriac pain and vomiting		Pancreatic head	5	Laparotomy, cystectomy, and cholecystectomy		Acute pancreatitis		
						Pancreatic body and tail						
		29	M				6	Laparotomy and cystectomy		None		
		25	F				9	Laparotomy, pancreatectomy and splenectomy		None		
Jai *et al* (2007)	1	26	M	Epigastric pain and pruritus	None	Pancreatic head	3	Laparotomy and partial cystectomy	None	None	Uneventful	(21)

Table I. Continued.

Authors (year of publication)	No. of cases	Age (years)	Sex (M/F)	Symptoms	PMH	Cyst location on the pancreas	Cyst size (cm)	Management	POAR	Complications	Follow-up	(Refs.)
Wu *et al* (2021)	1	28	F	Abdominal pain and jaundice	None	Pancreatic head	5.64	Laparotomy and pancreaticoduodenectomy	albendazole 800 mg/day	None	Uneventful	(22)
Soin *et al* (2019)	1	34	F	Epigastric pain	None	Pancreatic head	4	N/A	N/A	N/A	N/A	(23)

PMH, previous medical history; M, male; F, female; POAR, post-operative antibiotic regimen; N/A, not available.

of the cyst in the event of an equivocal diagnosis, enabling biochemical and cytological examination (13). In the case described herein, the imaging findings did not suggest a hydatid cyst, leading to the suspicion and consideration of a cystic neoplasm in the pancreas. Patients diagnosed with echinococcosis prior to surgery should receive prophylactic anthelminthic treatment with albendazole (10 mg/kg/day) for 2 to 4 weeks. This treatment should be continued for at least 4 weeks post-surgery to lower the risk of anaphylaxis (23). The patient in the present study did not receive anthelminthic treatment prior to the surgery as a diagnosis could not be made before the surgical procedure.

The EUS revealed a large, complex cystic-solid mass in the pancreatic body. A cytological examination of the fluid aspirated during the EUS revealed pyogenic inflammation with no evidence of malignancy. Based on the location of the hydatid cyst, various surgical techniques have been utilized for treatment, including pericystectomy, complete excision of the cyst, cysto-enteric anastomosis, distal pancreatectomy, capitonnage, open surgery and laparoscopy (7,17,19). In the present case report, the patient underwent a total pancreatectomy with partial excision of the attached second portion of the duodenum, as well as a splenectomy. He was administered a 4-week course of albendazole at a dose of 800 mg/day. During the surgery, utmost care should be exercised to mitigate the risk of contamination and subsequent recurrence. This involves minimizing the spillage of cyst contents through the application of scolicidal treatments, such as 0.5% cetrimide or 20% hypertonic saline (8). In the case in the present study, the entire pancreas was removed, and no chance was left for the intraoperative spillage of the cyst contents. A histopathological examination confirmed that the hydatid cyst caused chronic xanthogranulomatous.

A limitation of the present case report is the absence of data to definitively confirm that the death of the patient was due to diabetes complications.

In conclusion, pancreatic hydatid cysts are exceedingly rare and prone to misdiagnosis. Surgery remains the cornerstone of treatment, with a definitive diagnosis typically established through histopathological examinations.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

Authors' contributions

FHK and OHGH were major contributors to the conception of the study, as well as to the literature search for related studies. HAA and REA were involved in the literature review, study

design and writing of the manuscript. HHKA, DAE, DTG, KFHH, BAA and DHA were involved in the literature review, the design of the study, the critical revision of the manuscript, and in the processing of the table. FHK and BAA confirm the authenticity of all the raw data. RMA was the pathologist who performed the histopathological diagnosis. SHT was the radiologist who assessed the case. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent was obtained from the patient for participation in the present study.

Patient consent for publication

Written informed consent was obtained from the patient for the publication of the present case report and any accompanying images.

Competing interests

The authors declare that they have no competing interests.

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